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Year: 2010

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## **Lyme borreliosis presenting as hypersomnia**

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DOI: <https://doi.org/10.1007/s11818-010-0455-z>

Posted at the Zurich Open Repository and Archive, University of Zurich

ZORA URL: <https://doi.org/10.5167/uzh-43986>

Journal Article

Published Version

Originally published at:

Dalal, M A; Wehrle, R; Beitinger, P A; Wetter, T C (2010). Lyme borreliosis presenting as hypersomnia. *Somnologie*, (14):67-69.

DOI: <https://doi.org/10.1007/s11818-010-0455-z>

Somnologie 2010 · 14:67–69  
 DOI 10.1007/s11818-010-0455-z  
 Received: 3 November 2009  
 Accepted: 28 January 2010  
 Published online: 13 February 2010  
 © Springer-Verlag 2010

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# Lyme borreliosis presenting as hypersomnia

Idiopathic hypersomnia with long sleep time presents with a prolonged sleep episode of at least 10 hours, severe excessive daytime sleepiness, and the feeling of being unrefreshed or even somnolent upon awakening [1]. In Lyme encephalopathy, fatigue, memory problems, depression, and sleep disturbances may occur [2]. Although patients may complain about excessive daytime sleepiness (EDS), objective quantification of sleep in Lyme disease is rare. Polysomnographic measures of 11 patients with verified late Lyme disease revealed greater sleep latency and sleep fragmentation during nocturnal sleep compared to ten controls. No increased tendency to fall asleep during the daytime could be substantiated as mean sleep onset latency during the multiple sleep latency test (MSLT) was normal [3]. We report a patient who presented with isolated hypersomnia complaints, revealing Lyme borreliosis.

## Case report

A 22-year-old woman presented to our outpatient sleep clinic after having suffered from EDS over a 1 year period combined with intermittent involuntary sleep attacks lasting from seconds to minutes, occurring primarily during monotonous situations. Neither cataplexy, sleep paralysis, nor hypnagogic hallucinations were present. After work, she slept from 4:30 to 7:30 pm, had dinner, returned to bed by 8:00 pm and immediately fell asleep for 10 h until 6:00 am the following morning when she got up for work. On weekends, her sleep generally lasted 14–15 h after which she often woke up with a headache, feeling unrefreshed, and tired. Physical

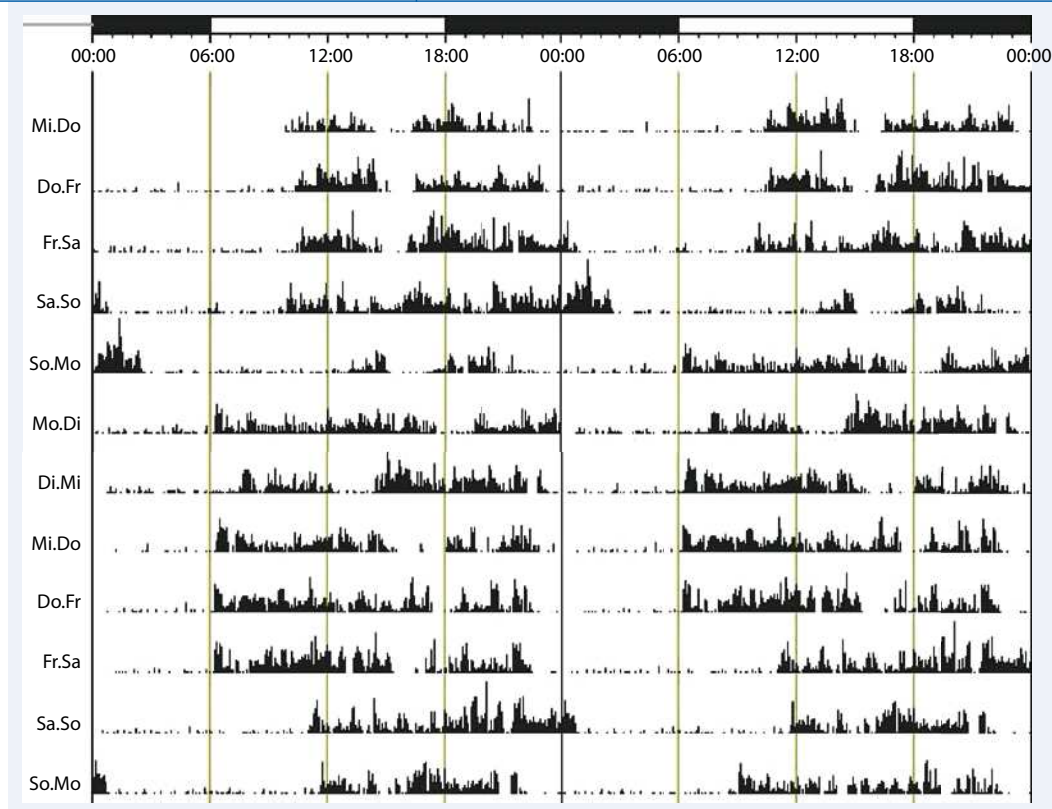
and neurological examination and electroencephalography (EEG) revealed no pathological results. Cranial MRI showed a small cyst of the pineal gland. The diagnostic work-up showed increased values for the inflammatory markers erythrocyte sedimentation rate (1 h: 13 mm/h) and the C-reactive protein (CRP) (7.28 mg/l; normal range 0.10–5.0 mg/l); other parameters, including complete blood count, creatinine, electrolytes, liver enzymes, bilirubin, urea, uric acid, lactate dehydrogenase, glucose, iron, ferritin, and basal thyroid stimulating hormone, were normal. Concerning prior clinical history, there were no indications for any viral illness, tick bites, or other skin lesions. Psychiatric

ly, the patient showed a slightly depressive mood and exhaustion. Actigraphic recordings over a 12-day period verified the excessive sleepiness with 9.4 h mean nighttime and 1.4 h mean daytime sleep, thus approximately 11 h of total sleep over a 24-hour period (■ Fig. 1). Polysomnography, including extended EEG recordings (F3, F4, Fz, T3, T4, T5, T6, Pz, O1, O2), and extended recording time (■ Tab. 1), reconfirmed the extended sleep period to 12.9 and to 9.4 h (with prolonged bed times of 14.3 and 11.2 h), without abnormalities in the sleep EEG or sleep architecture. The hypnogram of the recording of the first extended night is shown in ■ Fig. 2.

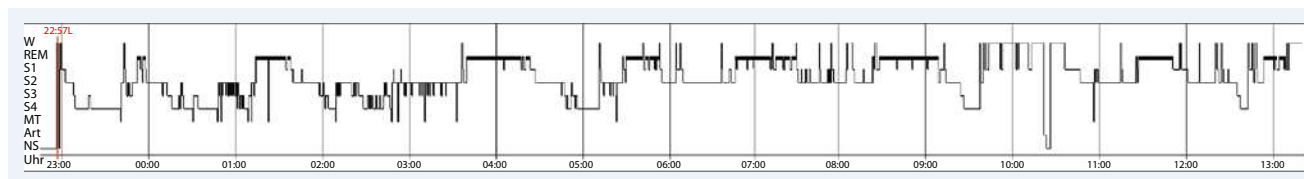
**Tab. 1** Measurements of polysomnographic recordings (PSG)

	Adaptation night 10:55 pm–6:57 am	First PSG (extended) 10:57 pm–1:20 pm	Second PSG (extended) 10:31 pm–9:42 am
Time in bed (min)	482.5	858.0	669.5
Total sleep time (min)	443.0	778.5	563.5
Sleep period time (min)	474.0	862.0	671.0
Sleep efficiency (%)	91.8	90.7	84.2
Sleep onset latency (min)	7.5	6.0	10.0
Slow wave sleep latency (min)	10.0	5.5	11.5
REM latency (min)	161.5	49.5	62.0
Wake time (% SPT)	6.2	7.4	13.0
Stage 1 sleep (% SPT)	0.6	10.0	9.0
Stage 2 sleep (% SPT)	48.2	47.6	33.5
SWS (% SPT)	27.4	19.9	20.5
REM (% SPT)	16.7	23.2	19.4
PLMS index	10.3	9.0	10.9
Apnea-hypopnea index	1.2	0.0	3.5
SaO <sub>2</sub> % mean	97.1	97.2	96.0
Arousal index	7.5	4.8	5.0

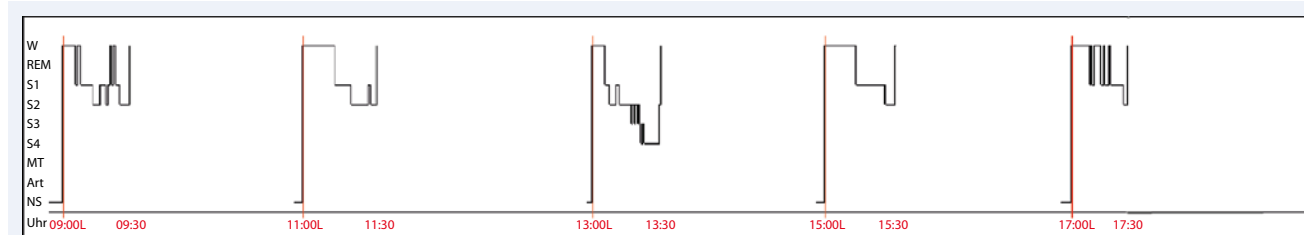
REM rapid eye movement, SPT sleep period time, SWS slow wave sleep (sleep stages 3+4), PLMS index number of periodic leg movements per hour of total sleep time, apnea-hypopnea index number of apneas and hypopneas per hour of total sleep time, SaO<sub>2</sub> blood oxygen saturation, arousal index number of arousals per hour of total sleep time.



**Fig. 1** ◀ Actigraphic recordings over a 12-day period verifying excessive sleepiness



**Fig. 2** ▲ Hypnogram of the recording of the first night showing an extended sleep period (for details see Table 1). Art Artifact, NS not classified, L lights off



**Fig. 3** ▲ Multiple sleep latency test (MSLT) revealing a shortened mean sleep latency (for details see text). L lights off

Absence of sleep-related breathing disorders or periodic leg movements (PLM) ruled out increased sleep pressure due to specific sleep-related disorders. Symptoms typical for restless legs syndrome (RLS) were denied. MSLT revealed that the patient fell asleep during each of the five tests with a shortened mean sleep latency of 5.5 minutes, once also reaching slow wave sleep (■ Fig. 3), thus reconfirming the subjective EDS (Epworth Sleepiness Scale score: 21/24). Lack of sleep-

onset REM periods and normal cerebrospinal fluid (CSF) orexin levels (239.7 pg/ml) ruled out narcolepsy. Lumbar puncture (LP), which was performed due to continuous poor cognitive performance and persistent headaches predominantly in the morning, revealed a neuroborreliosis with an increased borrelial-specific index for IgG antibodies (5.9) and a normal borrelial-specific index for IgM antibodies (<2.0). Borrelial antibodies were measured in the serum by Western Blot

and enzyme immunoassay: IgG revealed positive results, and IgM was marginally increased. In the CSF, IgG was positive, while IgM was not detectable.

## Discussion

In the present case with EDS, results of all diagnostic procedures according to the International Classification of Sleep Disorders (ICSD) standards including MSLT and extended sleep recording were con-

sistent with the suspected hypersomnia with long sleep time. LP, however, revealed neuroborreliosis as a possible cause for the symptoms reported. Following a 21-day intravenous antibiotic treatment with ceftriaxone (day 1: 1 g/day; days 2–21: 2 g/day), all symptoms disappeared almost completely. At two ambulatory follow-up examinations, the patient reported dramatic increases in subjective vigilance state, normalization of her sleep habits, and cessation of headaches. Our findings are limited in so far as we were not able to perform polysomnographic recordings including the MSLT following successful treatment in order to generate objective measures. Furthermore, false positive serologic tests can not be ultimately excluded.

The current diagnostic guidelines for hypersomnia mention inflammatory central nervous diseases as possible confounding diagnoses. Depending on the geographic location, Lyme borreliosis can be rather common, and it frequently overlaps with other neurological disorders [4]. Hence, LP should be considered in cases with suspected and polysomnographically verified hypersomnia, especially when additional headaches are reported, since the results may have important consequences regarding the treatment. In addition to psychiatric evaluation and brain imaging, LP may be included in the ICSD standards [1] as an additional diagnostic procedure to exclude inflammatory CNS disorders causing symptoms of EDS and abnormal sleep patterns [5].

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**Conflict of interest.** The corresponding author states that there are no conflicts of interest.

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## Abstract · Zusammenfassung

Somnologie 2010 · 14:67–69  
DOI 10.1007/s11818-010-0455-z  
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### Abstract

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### Keywords

Lyme borreliosis · Hypersomnia · Polysomnogram · Lyme disease · Lumbar puncture

## Hypersomnie als Hinweis auf Neuroborreliose

### Zusammenfassung

Hypersomnie geht häufig mit Symptomen wie Tagesschläfrigkeit, Kopfschmerzen und Abnahme der Vigilanz einher. Hier wird erstmals über eine Patientin berichtet, bei der Symptome wie bei einer idiopathischen Hypersomnie vorlagen, die sich aber nach genauer diagnostischer Abklärung als Neuroborreliose herausstellten. Dies unterstreicht die Notwendigkeit, ggf. auch diagnostische Maßnahmen wie eine Lumbalpunktion durchzuführen, um entzündliche Erkrankungen des Zentralnervensystems, welche die Symptome der Hypersomnie imitieren können, auszuschließen.

### Schlüsselwörter

Neuroborreliose · Hypersomnie · Polysomnographie · Lumbalpunktion